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A Tale of Two Diagnoses: An Interpretative Phenomenological Analysis of Healthcare Professionals Working with Children with Type 1 Diabetes and Coeliac Disease

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Abstract The psychosocial issues and challenges for young people with a dual diagnosis of type 1 diabetes and coeliac disease and their parents have been researched and uncertainties within screening and diagnostic processes are documented. However, the experiences of healthcare professionals working with this group have to date remained unexplored. A cross sectional design, using Interpretative Phenomenological Analysis, was used to understand the experiences of Health Care Professional's who work with children with a dual diagnosis and the meanings they assign to their experiences. Twelve healthcare professionals (paediatricians, gastroenterologists, nurses, dieticians) from three paediatric healthcare teams were interviewed about their experiences to explore the challenges, rewards and concerns in the role. Three superordinate themes emerged: 'connection and burden'; 'diagnoses together, but separate and uneven'; and 'sitting with certain uncertainty and complexity'. Themes represent participants' connection to the experience of patients and increasing patient burden; a sense of disparity in managing the two conditions; uncertainty and complexity with the dual diagnosis, and a repertoire of coping strategies utilised. Interpretations of participants' stories suggest that positive aspects of the role (i.e., making a difference, improving health and reducing risk) along with coping strategies serve to buffer the challenges of working with this client group. The study offers insight primarily into the challenges and coping responses of healthcare professionals working with the coeliac disease/ type 1 diabetes dual diagnoses. Recommendations include a dual review clinic (for type 1 diabetes and coeliac disease) and further support for staff. Additional research on psychosocial aspects of this dual diagnosis would support healthcare professionals in their role and service development.

Keywords: *healthcare professionals, experiences, children, type 1 diabetes, coeliac disease, dual diagnosis, interpretative phenomenological analysis*

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1. Introduction

With increasing emphasis on extending wellness and improving health, the Department of Health (DH) recognise that healthcare provision is becoming more complex with fresh challenges for professionals [1]. Complexity is pertinent to the area of long-term conditions, which are acknowledged to be challenging for healthcare professionals (HCPs), [2]. Caring for individuals with long-term health conditions requires a range of skills, which fall outside the traditional biomedical model of prevention, diagnosis and treatment [3]. Interventions are often rooted in patient self-management, which healthcare providers play a key

role in facilitating [4], however this role can be complex [5].

Exposure to on-going difficulties experienced by patients with long-term conditions may increase anxiety, feelings of professional inadequacy and emotional burden amongst clinical staff [6]. Burnout, a syndrome of emotional exhaustion [7] has been reported within healthcare professions. The additional energy required to provide compassion and empathy to individuals with chronic illnesses can have an emotional cost, which emphasises the importance of professionals' self-care [8]. Furthermore, to avoid burnout, it is important for clinicians to draw satisfaction from supporting patients in managing long-term conditions as opposed to being able to cure them [3].

A further challenge for staff in working with long-term conditions is the nature of uncertainty. The inability to abolish uncertainty in healthcare may create difficult challenges for clinicians and patients [9], amongst clinicians, treating individuals with multiple, coexisting health conditions has been associated with feelings of loss of control [10].

Studies discussing the experiences of HCPs caring for children with long-term illness highlight concerns about: sharing illness information [11]; the child and family's coping abilities [12] and capacity for illness self-management [13]. In addition, providing care within the complex relationships that can emerge between families and multiple services may create further challenges [14]. Two long-term conditions that are common during childhood are type 1 diabetes (T1D) and coeliac disease (CD).

The prevalence of CD in children with T1D is reported at between 5-7 times higher than the general population [15]. Children with a dual diagnosis of T1D and CD (hereafter referred to as dual diagnosis) usually present with asymptomatic or 'silent' CD [16]. Therefore, CD is likely to go undiagnosed unless children are screened [17]. The European Society for Paediatric Gastroenterology, Hepatology, and Nutrition (ESPGHAN) advise that children with an increased risk for CD should be screened [18]. Even if asymptomatic, those children testing positive are recommended to follow a GFD.

Psychosocial issues, including depression, anxiety, social withdrawal and lower quality of life, have been reported for young people with T1D [19,20,21] and emotional distress among parents [22,23,24]. Adherence difficulties, restrictions of activities and negative emotional responses (e.g., anger, feeling misunderstood) are documented for young people with CD [25,26]. Adhering to the GFD is in itself associated with psychosocial difficulties [27]. Thus, it seems plausible that the experience of managing the two conditions, a dual diagnosis, would magnify these challenges. The challenges faced by young people with a dual diagnosis and their parents has been explored in a qualitative study [28, unpublished data]. The nature of chronicity and variability with the diagnoses was linked to burden. A theme of protection and loss emerged, threatening future hopes and expectations for parents and adolescents, loss was associated with the adolescents' relationship with food and parental protection from future health complications.

It is unclear if CD and the GFD have implications for blood glucose control in T1D [29,30,31]. The evidence remains equivocal, thereby increasing the complexity of managing both conditions. Studies of the long term risks of untreated CD in children with T1D are also inconclusive, raising controversy for HCPs regarding optimum management [32,33]. The costs and benefits of screening and the additional burden of recommending a GFD (particularly to those asymptomatic for CD) have been discussed [33]. Challenges associated with the diagnostic process include the ethics of screening, requirement for sound information and support for patients and families [34].

Available literature provides evidence of the challenges for HCPs delivering care to those with chronic conditions, notably working with uncertainty, a lack of control and the need to promote self-management for patients. The challenges facing children and their parents associated

with T1D and CD include daily, life-long self-management, which can create physical and psychosocial complications that HCPs are required to identify and support. Therefore, besides the medical complications presented by this group, psychosocial issues add to the complexity for young people, their parents and HCPs [35]. Furthermore, the dual diagnosis can bring additional complexity associated with screening practices and recommendations. To date, the experiences of HCPs working with children and young people with a dual diagnosis remain unexplored. The current study explores the experiences of HCPs, working in multi-disciplinary teams with children and young people with a dual diagnosis, with the hope of furthering the understanding of this area of healthcare, potentially enhancing staff and service support.

2. Methodology

Interpretative Phenomenological Analysis (IPA) is a qualitative approach which explores how individuals make sense of their personal and social world [36]. IPA was chosen so as to gain an understanding of what it is like for HCPs to work with children and young people with a dual diagnosis and the meanings they assign to their experiences.

2.1. Participants

Participants (n=12; response rate 40%) were HCPs (of all healthcare disciplines) working in three multidisciplinary teams in the National Health Service (NHS) with children with a dual diagnosis of T1D and CD. Participants were required to have worked in this area for a minimum of three months (to allow them sufficient experience of working with a dual diagnosis to allow for meaningful reflection in interviews).

Table 1. Participant Demographics

Pseudonym	Profession	Team
Fran	Dietician	Diabetes
Charlotte	Dietician	Diabetes
Mandy	Dietician	Diabetes
Ann	Paediatrician	Diabetes
Sophie	Paediatrician	Diabetes
Kate	Consultant Paediatrician	Diabetes
Sam	Consultant Paediatrician	Diabetes
Ashley	Consultant Paediatrician	Diabetes
Clare	Specialist Diabetes Nurse	Diabetes
Rachel	Specialist Diabetes Nurse	Diabetes
Chris	Consultant Paediatric Gastroenterologist	Gastroenterology
Alex	Consultant Paediatric Gastroenterologist	Gastroenterology

Additional demographics (including age and number of years working in this area) were collected, however not reported here so as to protect participant anonymity.

2.2. Services

Due to the risk that some findings generated by the research could be attributed to influences of the service, participants were recruited from three services in separate

geographical locations across England and Scotland. All have paediatric diabetes teams working in close liaison with gastroenterology teams. Children and young people with a dual diagnosis were managed by their respective diabetes teams for both conditions across all participating sites, with additional input provided by the gastroenterologist in the case of further gastro-related complications, which is the usual service provision for these dual diagnosis patients. Participant demographics are shown in [Table 1](#).

2.3. Materials

Key themes from the literature on working with long-term illness were used to guide the development of initial questions, forming a list of open ended questions for the interview schedule. A dietician (not involved in this research) from the charity Coeliac UK, with experience of the dual diagnosis field, reviewed the interview schedule to assess the relevance and suitability of questions. In concordance with IPA principles, the interview schedule was used flexibly, to guide the exploration of experiences and meaning-making of participants, whilst following their concerns and ideas elicited during the interviews.

2.4. Procedure

Individual, semi-structured interviews (approximately 60 minutes) were conducted in participants' places of work and participants were debriefed after interviews. Each participant completed one interview.

2.5. Analysis

IPA data analysis by researcher (VH) followed the approach detailed by Smith et al. [37]. A brief outline of this approach includes:

- 1) Listening to interviews/ reading transcripts to note initial observations.
- 2) Line by line analysis of data (including descriptive, conceptual and linguistic details).
- 3) Developing emerging themes based on exploratory comments (including convergence and divergence).
- 4) Mapping connections between themes.
- 5) Putting aside these themes and connections in order to move on to the next transcript, analysing this independently (repeating steps 1-5 for each case).
- 6) Exploring connections or individuality between cases, assessing prominent themes, possible relabelling of themes.

Credibility was boosted by cross-checking coding with academic researchers (GUL and RH), who independently coded samples from three transcripts, which was then discussed with all researchers in a process of exploring concordance and disagreement of themes and interpretations. A sample transcript was additionally cross-checked by a University-based IPA analysis group. Throughout the analysis process, emerging themes were discussed in supervision and with the IPA analysis group. Resulting super-ordinate and subthemes were sent to the lead clinicians from each site for their views and as an opportunity for them to give feedback.

3. Results

Three superordinate themes, and eight subthemes, emerged from the data. A summary is provided in [Table 2](#). The results section will consider each theme, with quotations used for illustration. Attention will be paid to the general ideas found across participants, the nuances that were important to individuals and the connections between these.

Table 2. Summary of Themes

Superordinate Theme (Su.T)	Participants contributing to (Su.T)	Sub-theme (ST)	Participants contributing to (ST)
Connection and burden	All	Empathy and negative view of the diagnoses	All
		Feelings of adding to the burden (some guilt)	All
		Acting in the patients' best interests	Fran (D1), Kate (P), Clare (N), Chris (G), Ann (P), Ashley (P), Alex (G), Mandy (D), Rachel (N), Sophie (P)
Diagnoses together; but separate and uneven	10	Diabetes comes first and is often prioritised	Fran (D), Kate (P), Chris (G), Ann (P), Ashley (P), Charlotte (D), Mandy (D), Rachel (N), Sophie (P), Alex (G)
		Diagnoses going along in parallel but difficult to join up.	Fran (D), Kate (P), Chris (G), Ann (P), Charlotte (D), Mandy (D), Rachel (N),
Sitting with certain uncertainty and complexity	All	Complexity of the role	Clare (N), Chris, Kate (P), Charlotte (D), Rachel (N), Sophie (P)
		Diagnostic grey areas can feel difficult	Chris (G), Ann (P), Ashley (P), Charlotte, Alex (G), Mandy (D), Sophie (P)
		Striving for accurate control, sitting with sub-optimal control of the conditions	Fran (D), Kate (P), Clare, Chris (G), Ann (P), Ashley (P), Charlotte (D), Mandy (D), Sam (P), Rachel (N), Sophie (P)

3.1. Super-ordinate Theme 1: Connection and Burden

This superordinate theme features a strong connection from the HCPs to the experiences of the young people and their families. The empathy and connection perhaps heightens the challenges involved in making a diagnosis and supporting the management of the conditions. The enhanced feeling of responsibility for their role in this process seems to be managed well through a process of meaning making; seeing their role as one to keep the child healthy now and in the future. Experience in the role also seems beneficial in developing coping strategies.

3.1.1. Empathy and Negative View of the Diagnoses

The nature of having a dual diagnosis was frequently described negatively and as burdensome for young people; a sense of connection to the young person and family was found. All participants talked about the challenges of having a dual diagnosis. There was repetition of the notion “it’s bad enough having T1D, but to get CD too”. There were descriptions of the unfairness of the conditions, concerns about the impact they may have on the young person’s quality of life and a sense that it is harder for those diagnosed with CD experiencing few or no symptoms (asymptomatic). Empathy was demonstrated by participants positioning themselves in the shoes of the young person or parent. Depth of perspective taking may enhance the emotional connection HCPs have towards the patients and their experiences.

I think as a parent how hard that would be... I’ve always thought throughout my whole career, ‘thank goodness my child doesn’t have diabetes’. And then that added thought of having to deal with Coeliac disease on top is a massive challenge. Chris, Gastroenterologist; 28-30

However, there was also recognition of the range of responses found in families, with a feeling of admiration towards families that coped well with challenges.

3.1.2. Adding to the Burden

Generally, in children with a dual diagnosis, T1D is diagnosed first. Therefore, children are usually known to their diabetes team before receiving the CD diagnosis. Receiving a second lifelong diagnosis evoked compassion in participants. The language used to describe the second diagnosis (“the double whammy”, “a slap in the face”, “extra burden”) suggests how negatively (or realistically) they view this outcome.

I feel sorry... we know them because they’ve got diabetes so we’ve engaged with them, so you do feel, we feel sympathetic, so ‘oh, it’s the last thing they need’, you sit there and feel ‘ahh, what a shame’, Yeah you do feel that, you do. You feel ‘ahh, I didn’t want it for them’. Ashley, Paediatrician; 377-380

All participants shared the view that the second diagnosis created additional challenges and burden for young people and their families. Ashley’s excerpt (above) is reflective of having developed a relationship with the patient and family, connecting with them and reacting to the news of the second diagnosis, i.e. “I didn’t want it for them”. One participant gave an oppositional stance within

this theme, stating there is more to do practically with CD but that this is no different to managing T1D alone.

Almost half of the participants gave an account that portrayed a sense of responsibility, and in some cases guilt, for their role in ‘giving’ patients an extra set of issues to manage.

... when you throw in... Coeliac disease as well, you’re, you’re putting another pressure on the child.... you do feel bad kind of... having to lump something else on them ... you feel bad, feel awful for the child. Fran, Dietician; 83-102

Children with T1D who are asymptomatic for CD are picked up through the CD screening process. In these situations, the team diagnose a condition they see as burdensome and advise a GFD. This seems to be one of the challenges of working with the CD/T1D patient population, since these are situations where HCPs are not visibly bringing about improved health for the patient in the short-term (despite potentially improving health in the long-term), and perhaps elicit less gratitude from families.

3.1.3. Acting in Their Best Interests

The challenges described by participants were mostly followed up with descriptions of how they manage these feelings or make meaning from them. There is reference to responding differently with increased experience. Perhaps less guilt is experienced and a process of rationalising develops over time. Other nuances of this process of rationalising difficulties in their role included: knowing you have done as much as you can; following protocol; and acting in the child’s best interests. Participants described an important element of their role was to reduce risk; drawing this meaning from their role is perhaps a coping strategy, supporting them in bringing about a burdensome diagnosis and self-management regimes. The starkness of risk is highlighted in Alex’s quote.

...if you didn’t pick these patients up, if you didn’t screen them, what actually are the risks to the patient of gut cancer, other autoimmune conditions and bone issues or infertility... I actually don’t honestly know that we truly really know what the proper risks are..... but it’s not zero... Alex, Gastroenterologist; 209-220.

3.2. Superordinate Theme 2: Diagnoses are Together; but Separate and Uneven

This superordinate theme suggests there are varied views on prioritisation of the two conditions. Despite participants acknowledging the importance of CD, the dominant stance and tone was of T1D being prioritised by young people, families and the healthcare teams. Associated with this prioritisation, some professionals reported concerns about mixed messages being given to families, which can make their role more challenging. Barriers to providing equal care for both conditions included lack of time in clinic and lack of resources. The use of a joint clinic for the two conditions was reported to be helpful in demonstrating an equal weighting to families.

3.2.1. Diabetes Comes First and Is often Prioritised

Narratives comparing the conditions were provided by seven participants. The dominant view described T1D as more severe and risky in the short and long-term. Ashley portrayed an interesting analogy of quantifying the burden.

... if you look at the weighted burden of managing a gluten free diet versus the weighted burden of managing diabetes, the diabetes burden is far greater. I think if they were to be able to quantify those on a set of scales, the burden of diabetes is greater. Ashley, Paediatrician; 44-49

Participants spoke respectfully about the challenges experienced with CD, however there was an inclination towards describing T1D as the “worse” condition to have. This was linked to the possibility of reversing the symptoms of CD as long as the GFD is followed, and their view of the complications associated with both conditions. An opposing stance to this was depicted by Charlotte and Clare; they described how CD can be more arduous on a daily basis for families. Perhaps the HCP’s role is an influencing factor on this perception, with dieticians and nurses seeing different challenges based on the type of contact they have with families.

Participants described young people and families prioritising T1D over CD. This interpretation was linked to how families view the risks of T1D as higher, perhaps lacking the energy and resources to manage another condition or in some cases a lack of CD symptoms. This trend also emerged within the HCPs, with six reflecting a sense of priority towards the management of T1D or a perception that it is prioritised by others in the team. However, contrary to this, Mandy (dietician) was clear in her message of giving equality to both in order to treat the whole rather than the parts:

I don’t look at it as one or the other. For me they’re both equally as important, from a dietetic point of viewwe’re looking at it from the whole thing. Mandy, Dietician; 258-259

It seems reasonable to assume that diabetes teams would be inclined towards prioritising T1D based on it being the team focus, and considering the immediate, serious consequences of poorly controlled T1D. However, viewing the two conditions discretely perhaps creates barriers to working with the conditions more holistically.

3.2.2. Diagnoses Going along in Parallel but Difficult to Join up

Participants referred to factors that appear to create barriers to working with the conditions equally. Chris reflects a sense of responsibility to families to discuss their CD diagnosis, however, due to clinic resources, often cannot see them within a preferred time frame:

.....clinics are so full that they’re always too full to see children in a very timely manner and this is something that I’m conscious of, that by the time I’m often seeing them too long has gone by and I’m often erm feeling er that er, that we perhaps should’ve met up sooner... you might have already heard the diagnosis from the diabetic team. Chris, Gastroenterologist; 138-145

The phrases and tone used were indicative of deliberation, perhaps reflecting that the system is not ideal to fit the way Chris would like to support families. There appeared to be no additional time in appointments for dual diagnosis patients, which created a challenge to covering everything, more so if there were difficulties with one of the conditions. The importance of providing a consistent message to families about how to manage the conditions was indicated by half of the participants. The value of consistency from the team was linked to gaining trust and

the message it sends to families. Charlotte’s narrative suggests inconsistent messages can make her role difficult; she talks about less consistency with the team’s approach to CD, compared to T1D:

...always the first thing is getting the diabetes under control and everybody agrees and quite often it’s led from the doctors, they’ll be like ‘don’t worry about the gluten free diet’ but then that’s a difficult situation when later down the line you’re trying to then re-engage. Definitely for the Coeliac disease there isn’t as much of a team message, it tends to be more to the dietician that is constantly as the families would say ‘on at them’ whereas the doctors kind of leave it to us to deal with. Charlotte, Dietician; 522-537

There was also a feeling of hierarchy within these extracts; perhaps that families pay more attention to the advice given by the consultant. One of the three services offer a joint annual review, which was described as sending a subtle but positive message to the families about the importance of both conditions.

3.3. Superordinate Theme 3: Sitting with Certain Uncertainty and Complexity

Within this superordinate theme, professionals revealed the complexity that can exist within their roles; however they coped with this via processes of knowing their limits, being honest with families and seeking support. The rewards from “clear-cut cases” (symptomatic with clear CD diagnosis) seem to buffer the negative experiences of the “grey cases” (asymptomatic or unclear CD diagnosis). Another source of frustration was reported as coming from patient poor self-management, particularly when professionals lacked any understanding or reasoning for it. Despite an imbalance at times between professionals feeling they are putting a lot of effort in and making little difference, there is recognition of the need to accept sub-optimal control and maintain engagement.

3.3.1. Complexity of the Role

Five participants raised the issue of working with a condition that is not their area of specialty. This theme is illustrated by contributions from a diabetologist and gastroenterologist:

... being a diabetologist and not a gastroenterologist... sometimes we feel uncomfortable about them [diabetes patients with CD] and you’re not quite sure what exactly is the right thing to do. Sophie, Paediatrician; 104-108

... I do tend to hold back and not give as much information as I would do with children with just Coeliac disease. So I find that I’m probably not giving as much helpful information in an interaction... you’re avoiding mixed messages and saying the wrong thing. Chris, Gastroenterologist; 124-134

A lack of confidence was expressed by participants about working with the opposing condition to their specialist area; feelings of unhelpfulness and uncertainty were frequently described. Charlotte spoke of wearing two hats with dual diagnosis patients:

*... it’s remembering that through the consultation that they’ll ask you what about this? What about that? I find that quite difficult erm to remember to have both your hats on **all** the time with these patients and kind of remembering all aspects. Charlotte, Dietician; 285*

There was a tone here of needing to know or remember a lot and perhaps being asked questions outside her comfort zone. Charlotte commented on the quality of the service offered to dual diagnosis patients; she perhaps felt responsible for offering the same specific knowledge and service for CD as the patient would have if managed by the GI team. Kate provided a different stance on complexity below; she again puts onus on the dietician to have the specialist CD skills.

I think you probably don't need specialist or clinical skills to look after children with Coeliac disease, they need specialist dietetic skills but not doctor skills really. Kate, Paediatrician; 100-102

Participants described managing complexity by being honest with families about not having all of the answers and going to the 'expert' for advice (gastroenterologist or dietician). Having an expert to consult in such situations appeared to offer reassurance. The nature of responding to two conditions when you are a specialist in one appeared to add an element of complexity for some. There were extremes to this idea represented: for some it resulted in them feeling out of their comfort zones, less helpful or feeling that they were providing a lesser service. Others did not think that specialist skills were required or acknowledged that specialist skills were held by different professionals, with whom they could consult.

3.3.2. Diagnostic Grey Areas

There are uncertainties in the diagnosis, evidence base and management for the dual diagnosis and asymptomatic patients, described by half of participants.

...there always is an element of uncertainty, because a positive blood test doesn't equate to actually having the full blown picture of Coeliac disease. So er, they will sometimes go away with a degree of uncertainty and that causes anxiety... Chris, Gastroenterologist; 69-71

In circumstances where a diagnosis of CD is unclear, it seems that this brings trickiness for HCPs. Uncertainty in diagnosis can create anxiety in families, which might be a parallel process for HCPs too. Asymptomatic patients and those testing positive on the blood test (tTG) but negative on the endoscopy ('grey cases') seem the most challenging. Participants spoke about their uncertainty of not knowing what is the "right thing to do" with the grey cases. Circumstances where they were unable to give a definitive answer or were not able to see that they were making a positive difference might make management more challenging.

...on the one hand it's a very rewarding condition because it's a simple change of diet, albeit a big change for families, but you've made them better and you've reduced all those risks and they feel better. I think the more difficult ones are the patients that are asymptomatic. Alex, Gastroenterologist; 105-113

Five participants spoke of uncertainty in the evidence base for asymptomatic patients, with reference to the lack of good quality longitudinal research into the effects of not following a GFD for asymptomatic patients. The "best" treatment options for asymptomatic patients are described by Ashley as an "unknown":

... what really is the best treatment, and that's an unknown..... is there a benefit to glycaemic control to go on a gluten free diet? And that is very debatable. Because actually there may be evidence that actually, you exclude

the effect of having another diagnosis because they get exhausted by it all don't they, can't continue to carry all this burden, ... it's complex. Ashley, Paediatrician; 69-80

HCPs are trained to follow evidence-based practice; when this is not clear it perhaps makes it difficult to make recommendations. Ann's quote (below) is an example that once again, empathy is reflected by participants considering 'what would I do in that situation?'. And again, they manage this challenging position by being honest with the families about the facts, allowing them to make their decision about how they want to manage the condition, and turning to the "experts" for guidance.

...we all have discussions amongst ourselves, what would we do in that situation? ... you feel if all the people in their specialist fields have a degree of uncertainty then perhaps we don't know.... So yes, there is definitely some uncertainty about what to advise. Ann, Paediatrician; 316-328

In the grey cases, there seems to be an element of reassurance in the fact that the team has ongoing contact with the young person, and therefore they have the opportunity to detect health changes. There was also an element of acceptance from three of the consultants about the reality of medicine not always being certain.

3.3.3. Striving for Accurate Control but Accepting Sub-optimal

Participants acknowledged the range of patients they see. Nearly all participants referred to having families who responded very well and worked with the team; these were seen as the rewarding or easier cases. However, poor self-management was described as one of the challenges. Frustration when management of the conditions could be better was noted by eight participants. This was linked to concerns about health risk, and a sense of putting in a lot of effort but not seeing changes was described as "disheartening" (Sophie; 222).

It's hard, it's frustrating..... if the parents don't take it on board then the young person isn't going to take it on board...that's why I worry, that's what we're here for, to try and reduce you know the risks to future health. Rachel, Nurse; 227-142

The pivotal role of families in promoting effective management was highlighted by eight participants. A strong sense of empathy was shown for the challenges to families, however frustrations seemed to peak in cases where they cannot see why a family is unable to follow recommendations or "do the right thing", as shown in Charlotte's quote below. This perhaps reflects the complexity of psychosocial issues impeding adherence, which may be difficult to identify.

... it's very difficult.... we've got the child's best interests at heart and you are trying to support the families...there are plenty of other families that are in the same situation and they are managing to manage their diabetes and their gluten free diets. Charlotte, Dietician; 151-155.

There was a stance from participants of needing to accept when management of the conditions is not ideal. This was in acknowledgement of T1D and CD being difficult conditions to manage day-in and day-out. The idea of sub-optimal control being 'good enough' in some situations may be challenging to HCPs, who train from a medical model to alleviate symptoms and disease. It

seems that finding a balance between improving control of the diagnoses and engagement is a key aspect of the HCPs role, to improve outcomes from a clinical perspective, while having insight into the capacity of the young person and family, not “pushing them” to the point of losing them.

... just trying to keep that relationship going ... it's not perfect but you don't want to lose them, you want them to keep coming to clinic. Fran, Dietician; 410-416

There was also a theme running through the interviews of there being a limit to how much HCPs are able to do; after that it is the young people and families who choose whether or not they follow advice. This seemed to be another coping response to managing challenges of the role.

4. Discussion

The healthcare professionals' connection to the experiences of the young people with a dual diagnosis and their families (particularly their role in making the diagnosis) was important, and there was a felt sense of disparity in managing the two conditions. As with other areas of healthcare in the NHS, participants experienced uncertainty and complexity within their roles. The use of adaptive coping strategies and ways of meaning making were evident in order to combat difficulties in the presentation of the dual diagnosis, the lack of an evidence base and to manage how they feel.

Enhanced empathy and connection to patients' experiences was revealed in this study, which may reflect a process of building therapeutic relationships. Connection and compassion towards patients is cited as a wellness factor associated with the absence of burnout [38], and is also important to young people with long-term illnesses and their families [39]. Therefore, the findings of empathy and connection in this study are likely to be a positive influence on HCPs' wellbeing, the therapeutic relationship and patient care.

HCPs experienced a range of presentations working with a dual diagnosis, from “clear-cut cases” (symptomatic for CD with positive biopsy results) to “grey cases” (asymptomatic or those whose CD diagnosis is not certain following biopsy). In “clear-cut cases”, facilitating a diagnosis and health improvement were reported as highly rewarding for participants. However, in the “grey cases”, the sense of adding to patients' burden by screening and possibly finding an additional life-long diagnosis was less rewarding; this led to unease in most participants and feelings of guilt in some. The experiences told here support the findings by previous research [40] summarising that patient interactions for clinicians are a source of both satisfaction and emotional stress; the feeling of making a difference is important for physician wellbeing and can buffer stressful encounters. So within the range of dual diagnosis cases, circumstances where HCPs feel able to make a positive difference are likely to buffer the effects of the more challenging “grey cases”.

Almost all accounts of challenges were followed up with narratives that reflect meaning-making, coping, and appraising of roles and position in healthcare, including: promoting health; reducing health risks; following guidelines; and knowing they have done as much as they can. This process of cognitive reappraisal might serve as a

protective function and has been cited as a coping strategy in nurses [41]. The Process Model of Emotion Regulation (PMER) asserts that emotional impact is altered by changing how a situation is interpreted; reappraisal can decrease the experience of negative emotions and increase positive experience [42]. In this study, the HCPs' use of coping strategies to manage difficult experiences perhaps reflects professional hardiness and may serve to decrease the risk of burnout and emotional exhaustion.

Narratives and interpretations gave a sense of disconnection within the dual diagnosis, with a predominant view of T1D being prioritised. This finding is not surprising since it is the diabetes teams that also manage the CD in this study's sample, which is generalisable to management across the country. Interestingly, this theme was also found in a study of young people (and parents) with the dual diagnosis [28, unpublished data], where T1D was viewed as more threatening. It is possible that underlying messages of prioritising T1D could have clinical implications for present and future CD management and related physical and psychological well-being, such as not following the GFD or seeking support for self-management difficulties.

Professionals predominantly acknowledged the uncertainty within their roles; however, coping responses were varied. For some, thinking they have a lack of expertise in one of the conditions seems to have led to feelings of unhelpfulness, uncertainty and lack of confidence, which was found in both diabetes and CD specialists. It seemed that those who did not experience this difficulty were those who felt buffered by a feeling of security brought about by rationalising that it is not their role to be an expert in both (and they could consult other ‘experts’ for advice) and those who accepted that complexity and uncertainty are realities of modern day medicine.

A further source of complexity is working with families to facilitate optimal health management, and the frustrations when this is not ideal. Participants reflected how they strived for the best control for their patients (related to the reduction of risks), but were left with feelings of frustration when families who were deemed to have the resources needed to manage both conditions well, failed to do so. Psychological assessment and shared formulations are likely to have potential benefits (for professionals and patients) at these times. Again, coping strategies were used in response to poorer health management. Maintaining contact with families to monitor health, detect changes and keep them coming back to clinic were seen as important, along with being honest about areas of uncertainty, and equipping families with the facts to make their own decisions. Further responses involved an acceptance that “sub-optimal control” is sometimes a reality and a rationale that there is a limit to their capacity to bring about change.

5. Conclusions

Peer group reflective practice may boost the HCPs' support network in managing the dual CD/T1D patients. Regular continuous professional development events are recommended, to provide opportunities for specialists in both T1D and CD to share their expertise. Support in

formulating difficulties in treatment adherence may also be useful. An increased understanding of psychosocial issues (i.e., of patients presenting with reduced self-management) could help to reduce frustration for HCPs and to offer appropriate support for the young person and family. Despite participants indicating good multidisciplinary relationships and support, inconsistent messages between professionals in the team may increase challenges for some clinicians, particularly those who see their role as having more responsibility for managing one or other condition. Moving towards holistic management may include additional time in consultation, additional resources, and a dual review clinic.

References

- [1] Department of Health. (2008). High quality care for all: NHS Next Stage Review Final Report. London: The Stationery Office.
- [2] Royal College of Physicians, Royal College of General Practitioners and the NHS Alliance. (2004). *Clinicians, services and commissioning in chronic disease management in the NHS. The need for coordinated management programmes*. London: Royal College of Physicians.
- [3] Kane, R.L., Priester, R., & Totten, A.M. (2005). *Meeting the Challenge of Chronic Illness*. Baltimore: The John Hopkins University Press.
- [4] Schulman-Green, D., Jaser, S., Martin, F., Alonzo, A., Grey, M., McCorkle, R., Redeker, N. S., Reynolds, N. & Whittemore, R. (2012). Processes of self-management in chronic illness. *Journal of Nursing Scholarship*, 44, 136-144.
- [5] Thorne, S., & Paterson, B.L. (2001). Health care professional support for self-care management in chronic illness: Insights from diabetes research. *Patient Education and Counseling*, 42, 81-90.
- [6] Turner, J., & Kelly, B. (2000). Emotional dimensions of chronic disease. *Western Journal of Medicine*, 172, 124-128. Retrieved from <http://www.ncbi.nlm.nih.gov/pmc/articles/PMC1070773/?report=classic>
- [7] Maslach, C., & Jackson, S. (1981). The measurement of experienced burnout. *Journal of Organizational Behavior*, 2, 99-113.
- [8] Figley, C.R. (2002). Compassion fatigue: Psychotherapists' chronic lack of self-care. *Journal of Clinical Psychology*, 58, 1433-1441.
- [9] Han, P.K.J., Klein, W.M.P., & Arora, N.K. (2011). Varieties of uncertainty in health care: A conceptual taxonomy. *Medical Decision Making*, 31, 828-838.
- [10] Shapiro, J., Astin, J., Shapiro, S.L., Robitschek, D., & Shapiro, D.H. (2011). Coping with loss of control in the practice of medicine. *Families Systems and Health*, 29, 15-28.
- [11] Lochrie, A., Wysocki, T., Burnett, J., Buckloh, L.M., & Antal, H. (2009) Youth and parent education about diabetes complications: Health professional survey. *Pediatric Diabetes*, 10, 59-66.
- [12] Hoey, H., McGee, H.M., Fitzgerald, M., Mortensen, H.B., Hougaard, P., Lynggaard, H., Skovlund, S.E., Aanstoot, H.J., Chiarelli, F., Daneman, D., Danne, T., Dorchy, H., Garandeau, P., Greene, S., Holl, R., Kaprio, E., Kocova, M., Martul, P., Matsuura, N., Robertson, K., Schoenle, E., Sovik, O., Swift, P., Tsou, R.M., Vanelli, M., Aman, J. & Hvidøre study group on childhood diabetes. (2006). Parent and health professional perspectives in the management of adolescents with diabetes: Development of assessment instruments for international studies. *Quality of Life Research*, 15, 1033-1042.
- [13] Lake, A.J., & Staiger, P.K. (2010). Seeking the views of health professionals on translating chronic disease self-management models into practice. *Patient Education and Counseling*, 79, 62-68.
- [14] Dickinson, A.R., Smythe, E., & Spence, D. (2006). Within the web: the family-practitioner relationship in the context of chronic childhood illness. *Journal of Child Health Care*, 10, 309-325.
- [15] Sud, S., Marcon, M., Assor, E., Palmert, M.R., Daneman, D., & Mahmud, F.H. (2010). Celiac disease and pediatric type 1 diabetes: Diagnostic and treatment dilemmas. *International Journal of Pediatric Endocrinology*.
- [16] Telega, G., Bennet, T.R., & Werlin, S. (2008). Emerging new clinical patterns in the presentation of celiac disease. *Archives of Pediatric Adolescent Medicine*, 162, 164-168.
- [17] Saukkonen, T., Väisänen, S., Åkerblom, H.K., Savilahti, E., & the Childhood Diabetes in Finland Study Group. (2002). Coeliac disease in children and adolescents with type 1 diabetes: A study of growth, glycaemic control, and experiences of families. *Acta Paediatrica*, 91, 297-302.
- [18] Husby, S., Koletzko, S., Korponay-Szabó, I.R., Mearin, M.L., Phillips, A., Shamir, R., Troncone, R., Giersiepen, K., Branski, D., Catassi, C., Lelgemann, M., Maki, M., Ribes-Koninckx, C., Ventura, A., & Zimmer, K.P. (2012). European Society for Pediatric Gastroenterology, Hepatology, and Nutrition Guidelines for the Diagnosis of Coeliac Disease. *Journal of Pediatric Gastroenterology and Nutrition*, 54, 136-160.
- [19] Dantzer, C., Swendsen, J., Maurice-Tison, S., & Salamon, R. (2003). Anxiety and depression in juvenile diabetes: A critical review. *Clinical Psychology Review*, 23, 787-800.
- [20] Hanberger, L., Ludvigsson, J., & Nordfeldt, S. (2009). Health-related quality of life in intensively treated young patients with type 1 diabetes. *Pediatric Diabetes*, 10, 374, 381.
- [21] Weglage, J., Grenzebach, M., Pietsch, M., Eldmann, R.F., Innenbank, R.L., Denecke, J., & Koch, H.G. (2000). Behavioural and emotional problems in early-treated adolescents with phenylketonuria in comparison with diabetic patients and healthy controls. *Journal of Inherited Metabolic Disease*, 23, 487-496.
- [22] Bowes, S., Lowes, L., Warner, J., & Gregory, J.W. (2009). Chronic sorrow in parents of children with type 1 diabetes. *Journal of Advanced Nursing*, 65, 992-1000.
- [23] Haugstvedt, A., Wentzel-Larsen, T., Rokne, B., & Graue, M. (2011). Perceived family burden and emotional distress: Similarities and differences between mothers and fathers of children with type 1 diabetes in a population-based study. *Pediatric Diabetes*, 12, 107-114.
- [24] Urquhart Law, G., Walsh, J., Queralt, V., & Nouwen, A. (2013). Adolescent and parent diabetes distress in type 1 diabetes: The role of self-efficacy, perceived consequences, family responsibility and adolescent-parent discrepancies. *Journal of Psychosomatic Research*, 74, 334-339.
- [25] Altobelli, E., Paduano, R., Gentile, T., Caloisi, C., Marziliano, C., Necozone, S., & di Orio, F. (2013). Health-related quality of life in children and adolescents with celiac disease: Survey of a population from central Italy. *Health and Quality of Life Outcomes*, 11, 204-213.
- [26] Roma, E., Roubani, A., Kolias, E., Panayiotou, J., Zellos, A., & Syriopoulou, V.P. (2010). Dietary compliance and life style of children with coeliac disease. *Journal of Human Nutrition and Dietetics*, 23, 176-182.
- [27] Howard, R., & Urquhart Law, G. (2014). Psychological well-being of children and young people with coeliac disease: management and intervention. In C. Martin & T. Dovey (Eds.), *Paediatric Gastrointestinal Disorders: A psychosocial perspective* (pp. 186-207). London: Radcliffe Publishing Ltd.
- [28] Love, M.E. (2013). *Living with coeliac disease and type 1 diabetes: The experiences of adolescents and their parents*. Psy.D thesis, University of Birmingham: UK.
- [29] Mohn, A., Cerruto, M., Iafusco, D., Prisco, F., Tumini, S., Stoppoloni, O., & Chiarelli, F. (2001). Celiac disease in children and adolescents with type 1 diabetes: Importance of hypoglycemia. *Journal of Pediatric Gastroenterology & Nutrition*, 32, 37-40.
- [30] Saadah, O.I., Zacharin, M., O'Callaghan, A., Oliver, M.R., & Catto-Smith, A.G. (2004). Effect of gluten-free diet and adherence on growth and diabetic control in diabetics with coeliac disease. *Archives of Disease in Childhood*, 89, 871-876.
- [31] Schwarzenberg, S.J., & Brunzell, C. (2002). Type 1 diabetes and celiac disease: Overview and medical nutrition therapy. *Diabetes Spectrum*, 15, 197-201.
- [32] Freemark, M., & Levitsky, L.L. (2003). Screening for celiac disease in children with type 1 diabetes two views of the controversy. *Diabetes Care*, 26, 1932-1939.
- [33] Sud, S., Marcon, M., Assor, E., Daneman, D., & Mahmud, F.H. (2011). Quality of life in children with diabetes and celiac disease: Minimal impact of the 'double diagnosis'. *Pediatric Diabetes*, 13, 163-169.
- [34] Mimmagh, J. & Thornton, H. (2006). Coeliac disease and type 1 diabetes in a paediatric setting. *Journal of Diabetes Nursing*, 10, 222-226.

- [35] Howard, R., Urquhart-Law, G., & Petty, J. (2012). Coeliac disease: Psychosocial factors in adults and children. In C. Lloyd & T. Heller (Eds.). *Long-term Conditions: Challenges in Health and Social Care*. (pp. 108-124). London: SAGE.
- [36] Smith, J.A. & Osborn, M. (2008) Interpretative phenomenological analysis. In J.A. Smith (Ed.) *Qualitative Psychology: A practical guide to research methods* (pp. 53-80). London: Sage.
- [37] Smith, J., Flowers, P., & Larkin, M. (2009). *Interpretative Phenomenological Analysis: Theory, Method and Research*. London: SAGE.
- [38] Eckleberry-Hunt, J., Lick, D., Boura, J., Hunt, R., Balasubramaniam, M., Mulhem, E., & Fisher, C. (2009). An exploratory study of resident burnout and wellness. *Academic Medicine*, 84, 269-277.
- [39] Taylor, R.M., Gibson, F., & Franck, L.S. (2008). The experience of living with a chronic illness during adolescence: A critical review of the literature. *Journal of Clinical Nursing*, 17, 3083-3091.
- [40] Wallace, J.E., & Lemaire, J. (2007). On physician well being—you'll get by with a little help from your friends. *Social Science & Medicine*, 64, 2565-2577.
- [41] Lambert, V.A., Lambert, C.E., Petrini, M., Mei Li, X., & Zhang, Y.J. (2007). Workplace and personal factors associated with physical and mental health in hospital nurses in China. *Nursing and Health Sciences*, 9, 120-126.
- [42] Gross, J.J. (2002). Emotion regulation: Affective, cognitive, and social consequences. *Psychophysiology*, 39, 281-291.
- [43] White, L.E., Merrick, V.M., Bannerman, E., Russell, R.K., Basude, D., Henderson, P., Wilson, D.C., & Gillett, P.M. (2013). The rising incidence of celiac disease in Scotland. *Pediatrics*, 132, 924-931.
- [44] Patterson, C.C., Gyurus, E., Rosenbauer, J., Cinek, O., Neu, A., Schober, E., Parslow, R.C., Joner, G., Svensson, J., Castell, C., Bingley, P.J., Schoenle, E., Jarosz-Chobot, P., Urbonaite, B., Rothe, U., Krzysnik, C., Ionescu-Tirgoviste, C., Weets, I., Kocova, M., Stipancic, G., Samardzic, M., de Beaufort, C. E., Green, A., Dahlquist, G.G., & Soltesz, G. (2012). Trends in childhood type 1 diabetes incidence in Europe during 1989-2008: evidence of non-uniformity over time in rates of increase. *Diabetologia*, 55, 2142-2147.